Scientiometric Review on Zika and its Relationship with Craneofacial and Oral Disorders

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Abstract

The Zika virus (VZ) is a neurotropic virus that causes illnesses in the unborn baby, when a pregnant mother is infected. Given the relationship that exists between this virus and craniofacial and oral alterations, it is necessary to review high-impact information to clarify the consequences that it left in this child population.

Objective: Review articles in databases during the years 2016 to 2020 with the subject in relation.

Methods: A bibliometric analysis was carried out with the research articles found in the WOS + Medline + Scielo databases in the years 2016 to 2020, regarding the issue of VZ and its relationship with craniofacial alterations, in which it was had as a result the analysis of 18 articles, reviewing the inclusion of terms in the title, abstract and keywords.

Results: The co-occurrence has a higher incidence in the term of Microcephaly, other results found is the analysis of the countries with the largest publications on the subject is Brazil and taking into account the year of publication from 2016 as the date of the disease outbreak, 2019 was the year with the highest number of publications.

Conclusions: The study concluded that according to the theme and craniofacial alterations, there is very little scientific evidence that relates the two terms and among Latin American countries a report was found in the countries of Brazil, Canada and the United States.

Key words:

Zika Virus; Microcephaly; Skull; Face; Oral Manifestations.

Introduction

In 2015, the Zika epidemic arrived in Colombia, during which 99,721 cases were reported, of which 8,826 were laboratory-confirmed and 90,895 were clinically suspected. This is how in 2018 at least 318 children born with microcephaly were reported in the country (***Candelo et al., 2018***).

The reported prevalences of microcephaly from 2007-to 2011 show a prevalence of 4.83 evidencing in 2015 a prevalence of 1.66. In 2016 at 8.96, in 2017 at 5.95, and the first half of 2018 at 3.85. This evidences a real increase in 2016, 2017, and 2018 (***Candelo et al., 2018***).

Zika is a virus with a strong neurotropic effect that is spread by an Aedes vector. Its contagion determines the appearance of an outbreak of skin rash in the pregnant mother that triggers several signs, the most evident being microcephaly. However, infections in advanced pregnancies, and babies with normal skull sizes, can also present significant brain damage (***França et al., 2016***).

Before the ZIKA epidemic, microcephaly ranged from 0.3 to 3.1 per 10,000 births, with an average of 1.8 (95/100 CI 1.7-1.8) (***Fonteles et al., 2018***). For the same period, 381 neurological syndromes were reported, of which 258 are Zika-associated Guillain Barré syndromes(***França et al., 2016***)(***Candelo et al., 2018***). Microcephaly affects cranial bone arrangement, facial growth, and neurological syndrome affects muscle tone and the process of chewing, swallowing, and airway arrangement, which has not yet been characterized. Other studies report multiple systemic alterations associated with neurological failures, such as facial paralysis, dysphagia, and muscular hypotonia (***França et al., 2016***).

The World Health Organization (WHO) in July 2018, refers that VZ in transmission from pregnancy causes microcephaly and other malformations, which constitute the congenital syndrome due to VZ. This syndrome includes other malformations such as contracture of the limbs, muscular hypertonia, ocular alterations, and deafness. Similarly, it is estimated that 5 to 15/100 of infants born to women infected during pregnancy present complications associated with the virus (***Candelo et al., 2018***).

Given the above, the present research aims to address the subject from the dental point of view and proposes the following research question: What is the tendency of publication of bibliographic records in indexed sources in the Web of Science main collection, Medline, and Scielo databases on the subject of Zika associated with craniofacial and oral alterations?

Until the beginning of this study, no information was found on VZ and its relation to the oral cavity, so the need arises to analyze the bibliometric trend in this subject. In addition, the databases consulted offer the greatest coverage of refereed scientific journals in the areas of health sciences, both in the global context and in the context of Hispanic America (***Martín-Martín et al., 2021***).

Since 2017, there is a consensus in reference that VZ, generates microcephaly, and is associated with indicators of Guillan-Barre Syndrome, as well as neurological disorders and seizures (***Fonteles et al., 2018***). This virus has not defined the systems it alters and the medium and long-term consequences it entails. There is minimal evidence describing the facial and intraoral characteristics of children affected with ZIKA, Brazil described alterations in the lingual frenulum (***Dain Gandelman Horovitz et al., 2016***), and another study describes that there are no specific oral manifestations associated with ZIKA, however, most studies describe the neurological characteristics related to the syndrome, being more evident the microcephaly (***Garcez et al., 2016***).

On the other hand, other less clear situations establish how these defects will affect the survival and quality of life of those affected (***Leão et al., 2017***). Derived from the above, the need arises to identify from publications in peer-reviewed scientific journals, and published literature on the issue of oral manifestations of Zika. To identify who has conducted research in this field, where they have worked (institutions and countries), and in which journals such publications have been made.

At the same time, it is expected that the results will be inputs to transcend in this research with a multidisciplinary team, based on the review of the quality of the scientific evidence available and that will contribute to improve the living conditions of this population.

Given the above, the purpose of this study was to perform a bibliometric analysis of the research articles indexed in the Web of Science + Medline + Scielo databases from 2016 to 2020, regarding the topic of VZ and its relationship with craniofacial alterations. Likewise, the countries in which more publications were made concerning the studied topic were defined and the years in which the publications made about said topic predominated were reviewed.

Methods & Materials

Population

This research was developed at Santo Tomás University, with a work team belonging to the Faculty of Dentistry, since March 2020, advised by the staff and bibliometric tools available to The Resource Center for Learning and Research CRAI, of the same university.

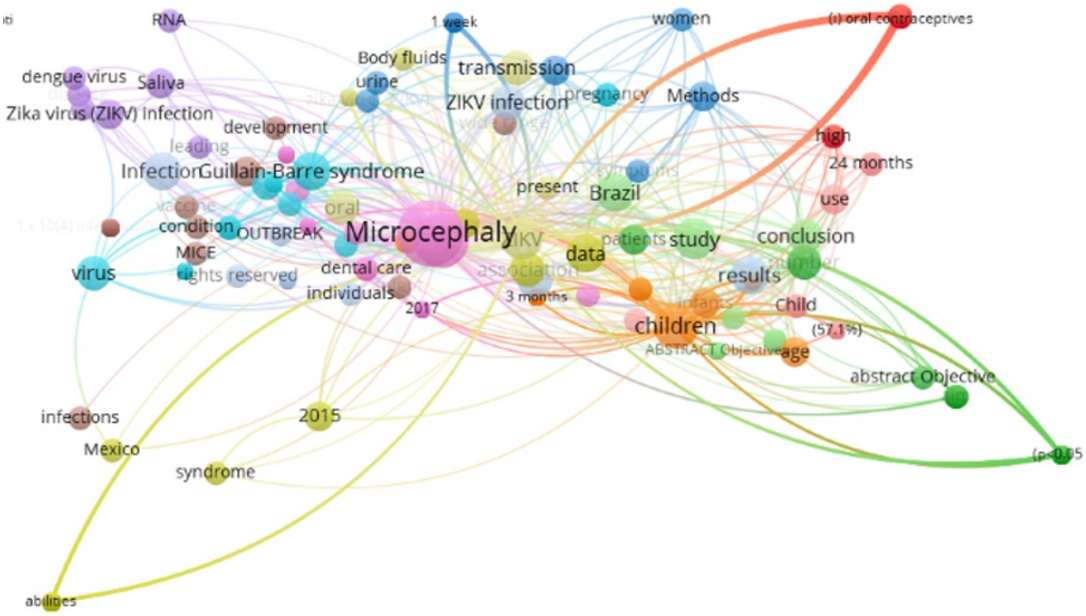

Figure 1. Map of Keywords.

Maplike representation of research topics included in the titles, abstracts, and keywords

A bibliometric analysis was carried out with the research articles found in the WOS+Medline+Scielo databases in the years from 2016 to 2020, which is observational, descriptive, and cross-sectional. On the other hand, it is a retrospective study by reviewing publications in the past period in journals indexed in bibliographic databases.

Ethical considerations were not taken into account for this study, due to the type of study, which does not require direct contact with patients. The articles that included in their title, abstract, and keywords the terminology between Zika and alterations in the skull and face were selected, this being the main search variable. Articles with the term Zika but not associated with craniofacial and oral alterations were discarded. Other variables related to the results were year and country of publication.

Statistical analysis

For the search of the articles, the following search equation was defined TS=((zika OR "Congenital zika syndrome" OR "ZVD") AND Microcephaly AND ("Maxillofacial, clinical characteristics" OR oral)) based on the keywords of MESH terms.

We proceeded to search the following databases Web of Science main collection (15 records) + Medline (15 records) + SciElo (6) independently, processed in VantagePoint. Activity indicators were obtained based on publication frequency by years, countries, and authors. In addition, the co-occurrence of research concepts in titles, abstracts, and keywords was analyzed. The terms were grouped according to the categories of multiword phrases, keywords-author, phrases-title, phrases-abstract, keywords-plus, dentistry, and microcephaly.

From the previous combination without duplicates in the information, 18 articles were found, also taking into account the years of publication and the countries where they were published. Subsequently, the information from the co-occurrence matrix of key terms was taken to the VOSviewer, and the interrelation of the variables was observed using a co-occurrence map. The sample size was the totality of the articles found.

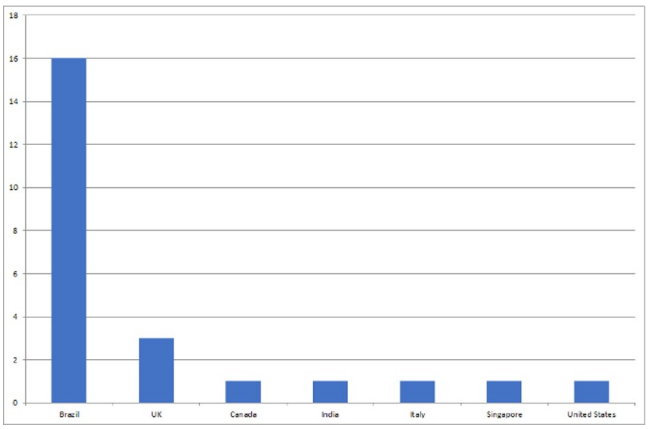

Figure 2. Associated countries Countries and territories by number of scientific publications on Zika virus related to craniofacial alterations

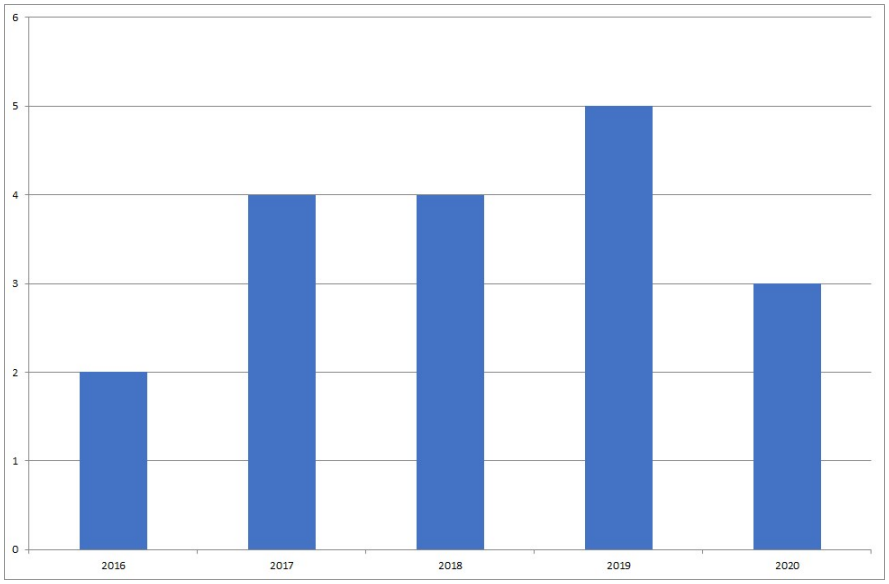

Figure 3. Years of publication Years of publication, of scientific papers on Zika virus related to craniofacial alterations

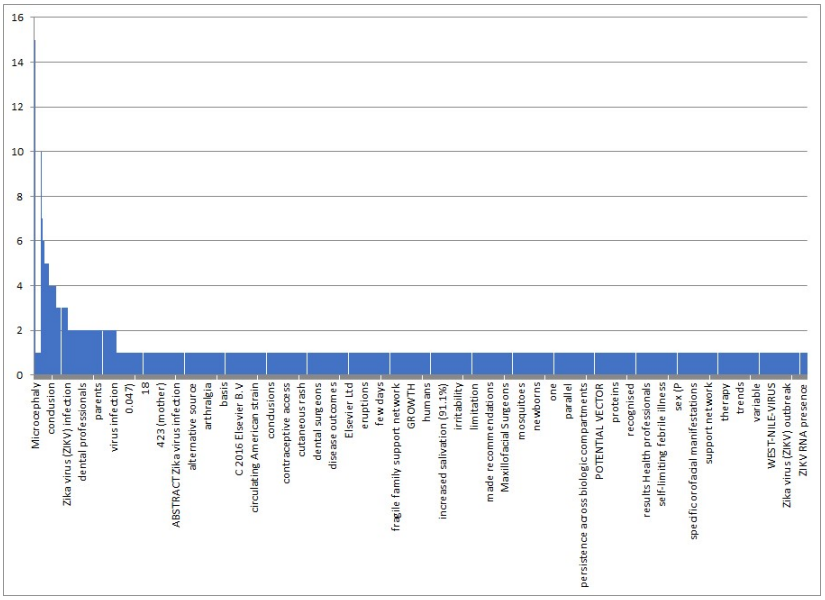

Figure 4. Keyword graph keywords identification used in the publication trend

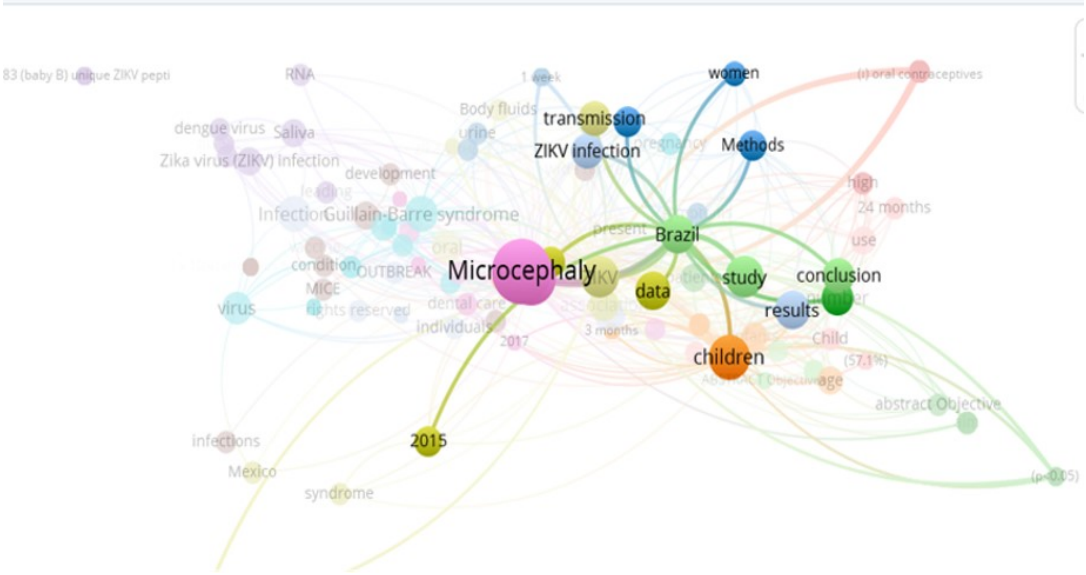

Figure 5. Keyword mapping sizes of the nodes related to the number of articles associated

Results

Taking into account the search equation, 839 articles were defined, of which the articles with the most records were taken, leaving 18 articles, of which 8 were discarded. Subsequently, with the citation analysis, 8 more articles are found. The figure 1 shows the map of co-occurrence of keywords that allows locating in a map-like representation the various research topics included in the titles, abstracts, and keywords describing the articles (***Saad et al., 2018***). The size of the nodes is associated with the number of publications in which the terms appear and the location on the map indicates that the terms tend to appear more frequently together concerning others at opposite ends (Figure 1). In addition, this graph showed an interrelation between the 839 keywords, where a higher incidence of the term Microcephaly was evidenced, and the presence of other terms such as children, Zika Virus, Brazil, outbreak, Saliva, Dental care, body fluids, transmission, pregnancy, Zika infection. The review was also carried out by the countries of affiliation of the publications and as a result, the record of publications of the countries associated with the subject studied was found, as can be seen in Figure 2.

This bibliometric study showed that the country that published the most articles on Zika virus related to craniofacial alterations was Brazil with 16 publications. This was followed by the United Kingdom with 3 publications. Finally, Canada, India, Italy, Singapore, and the United States followed, all with 1 publication (Figure 3). It is highlighted that the predominant publications were made by authors in Latin American institutions, due to the contributions made by Brazil. This country is one of the most affected by this disease. Another bibliometric data are the years of publication as evidenced in Figure 4, which takes into account the epidemiological outbreak of this disease, the bibliometric search was performed from 2016 to date, finding reports in the years 2016 continuously until 2020, taking only the articles published in Latin America. The years with the highest number of publications were 2019 with 5 publications and 2017 and 2018 with 4 publications, which would correspond to 72.2 percent. In the range of years studied, the year with the lowest number of publications was 2016 with 2 publications and the year with the highest number of publications was 2019. Similarly, publications were included in 2020, up to June, corresponding to 3 publications. The year with the highest number of publications in the associated subject was 2019 with 5 publications.

Discussion

No bibliometric reviews were found based on this topic, so it was not possible to make a relevant comparison of the present study. Due to this, it is necessary to carry out further research about VZ and its relationship with craniofacial alterations.The keywords allow identification of the publication trend, as shown in Figure 5. The sizes of the nodes are related to the number of articles associated with them. The larger the nodes, the greater the number of associated publications.

Likewise, the closeness of the nodes indicates the number of times the terms appear together and, on the contrary, if they appear at opposite ends, the words do not tend to appear together in the publications studied. In the present study, it was observed that the largest node was Microcephaly, indicating its relevance in terms of publications, this result coincides with several articles, one of the studies describes five specific characteristics of this syndrome and one of them is severe Microcephaly with partially collapsed skull (***Moore et al., 2017***), another study, according to WHO states that based on the review of observational, cohort and case-control studies, there is a scientific consensus that VZ is a cause of microcephaly and other neurological complications that constitute a congenital syndrome (***Siqueira et al., 2018***).

In Figure 6, another result is the publication according to the affiliation of the countries, recognizing that Brazil is the country with more publications, highlighting that, of the 18 articles, 8 were made on the population of different states in Brazil, other important nodes are Children, Brazil, Outbreak (***Fonteles et al., 2018***; ***Saad et al., 2018***; ***Satterfield-Nash et al., 2017***).

The bibliometric study also identified neurological alterations, as referred to in one of the review

articles (15), which analyzed the vertical relationship of VZ infection, describing a wide spectrum of neurological alterations with and without microcephaly, some of them asymptomatic, which can lead to neuropsychomotor delays, epilepsy, and visual anomalies; in coincidence with our objective, this study aimed to study the natural history of the disease to promote better development and quality of life. identifying that, within the alterations in newborns, there are exacerbated primitive reflexes that disappear later than expected. Another study conducted between 2015 and 2016 in 15 Brazilian states that presented laboratory-confirmed VZ transmission and due to the increase of microcephaly in newborns, evaluated 19 children aged 19 to 24 months, who presented motor disability, seizure disorders, and auditory and visual abnormalities.

Although the above diagnoses are described in early infancy, later development has not yet been characterized, which limits treatment planning and support for the child and family members ((***Satterfield-Nash et al., 2017***)). At the dental level, a study evaluated the lingual frenulum in children affected with congenital zika syndrome and the association of lingual frenulum phenotypes with other variables (***Fonteles et al., 2018***).

This article coincides with the neurological diagnoses and also with the lack of functional and structural diagnoses at the intraoral level and the influence of these aspects on the development of the stomatognathic system functions. Among the results, they observed many infants affected by congenital zika syndrome (CZS), which presented a posteriorly positioned lingual frenulum, instead of the absence of this small band of tissue the lingual frenulum of 34 infants, and in 14 of them, the frenulum was identified as submucous.

Another comparative study, performed in northeastern Brazil, analyzed the alterations in primary teeth of 108 children with and without microcephaly and aged 0 to 2 years attending public services, although the study does not express the definitive diagnosis associated with VZ, the patients were studied in the period of the highest epidemiological peak in Brazil between 2015 and 2017, where it was declared as an International public health emergency, in its results, it describes that children with microcephaly present greater delays in chronology, alterations in the sequence of eruption and enamel defects unlike children who did not present ((***Saad et al., 2018***)); In addition, he reports that the tropism of this virus in the nervous tissue can cause serious repercussions to the baby. These results are very close to what we intend to evaluate in this study, recognizing that this subject requires further studies to evaluate the dental alterations that occur in patients with congenital VZ syndrome.

Author contributions

Conceptualization: MJAM; Methodology: MJAM; Software: ASCC; Validation: CAAA; Formal analysis: MJAM; Investigation: ASCC; Resources: MJAM; Writing - original draft: ASCC; Writing - review & editing: ASCC, CAAA; Visualization: MJAM

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